Title

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CARDIOVASCULAR IMAGES

Using a 3-Dimensional Printed Model to Plan Percutaneous Closure of an Unroofed Coronary Sinus

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A 61-year-old man diagnosed with pulmonary arterial hypertension 5 years ago treated with tadalafil, atrial flutter status postablation, and multiple myeloma status post bone marrow transplantation 1 year prior presented for evaluation of exertional dyspnea. Physical examination revealed jugular venous distension and a mid-systolic murmur heard at the left upper sternal border without radiation.

A transthoracic echocardiogram was notable for mild right ventricular enlargement with normal biventricular systolic function, no significant valvular abnormalities, estimated pulmonary artery systolic pressure of 35 mm Hg and evidence of interatrial shunting after agitated saline administration. Transesophageal echocardiography (TEE; Figure 1A and 1B) and cardiac computed tomography angiography (Figure 1C) revealed a fully unroofed coronary sinus (CS) defect that extended over the entire left atrial course of the CS and a dilated main pulmonary artery measuring 40 mm. No anomalous pulmonary venous return or persistent left superior vena cava were seen.

Given the patient's impaired functional capacity and right ventricular enlargement, the decision was made to proceed with unroofed CS defect closure. The patient strongly preferred to avoid open-heart surgery, and hence percutaneous device closure was considered. Given the unique abnormality, a 3-dimensional (3D) model was printed to assess for feasibility of percutaneous closure. Cardiac computed tomography angiography scan DICOM files were loaded as a series into ITK-SNAP1 software (University of Pennsylvania, Philadelphia, PA) for segmentation of the contrast-enhanced blood pool, creating a 3D stereolithography file. A 1.5 mm shell was formed around the solid blood pool model using Autodesk Meshmixer (Autodesk, San Rafael, CA) to create a hollow model representing the inner, endocardial surface of the heart. The hollow stereolithography model was 3D printed on a Formlabs Form 3 SLA printer (Formlabs, Somerville, MA) using solid resin (Figure 1D and 1E).

We then performed bench testing of various percutaneous closure devices using the 3D model, positioning the left atrial disk in the left atrium and the right atrial disk within the CS in its right atrial aspect. We deemed that percutaneous closure would be feasible with either a 44 mm GORE Cardioform ASD occluder device (Gore Inc, Newark, DE) or a 30 mm Amplatzer septal occluder (Abbott, Plymouth, MN; Figure 1F).

A baseline congenital cardiac catheterization was performed, with results shown in the Table. Given the large shunt fraction of 2.5:1 and normal pulmonary vascular resistance, we proceeded with device closure. Real-time TEE was used for interventional guidance. Balloon sizing of the defect with a 34 mm AGA sizing balloon revealed a stop-flow measurement of 25 mm by TEE. Based on this measurement and results of bench testing with our 3D model, a 44 mm GORE Cardioform ASD occluder device was positioned in the inferomedial aspect of the

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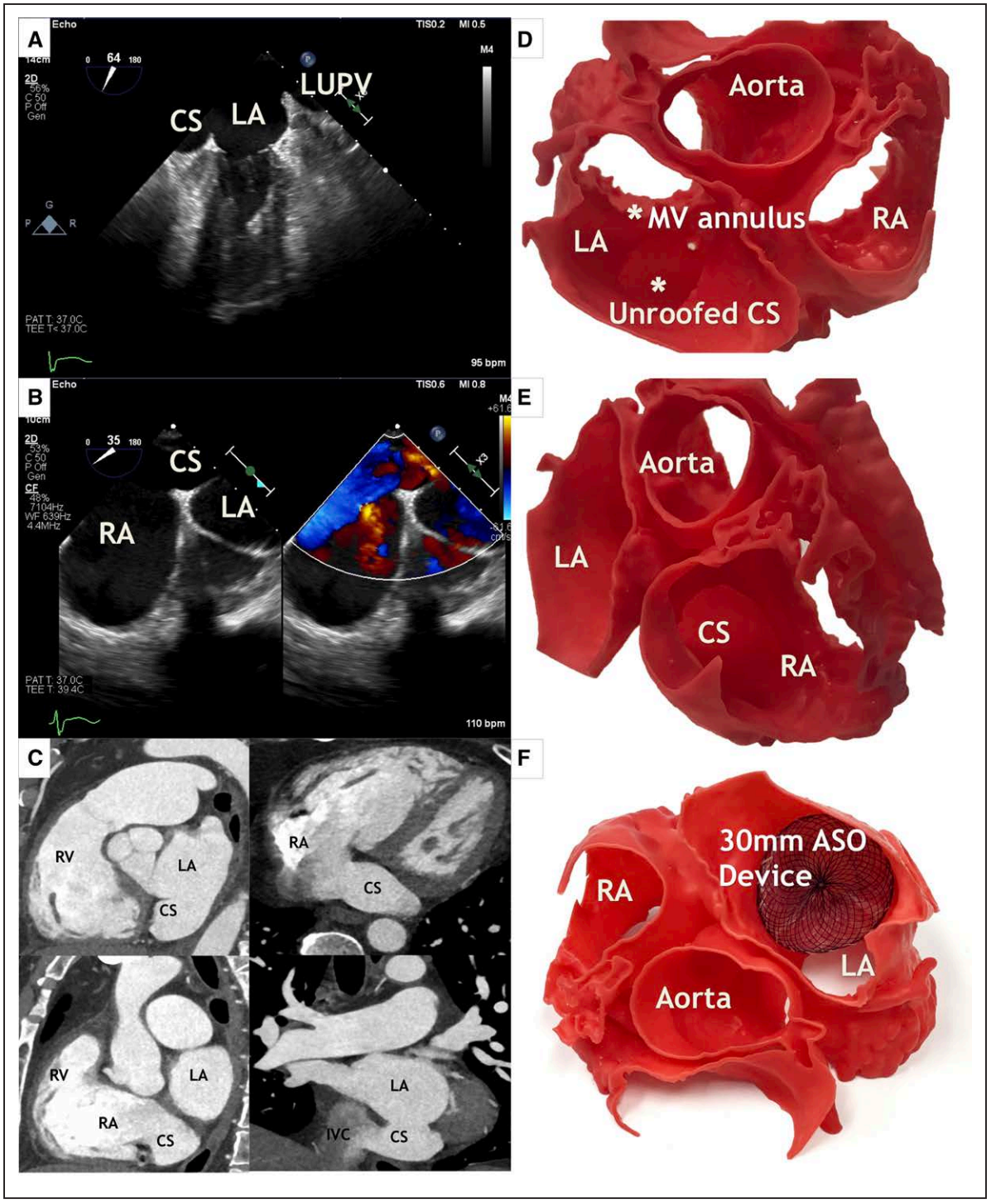

Figure 1. Multimodality imaging of unroofed coronary sinus (CS) atrial septal defect.
After diagnosis was made by transesophageal echocardiography (**A** and **B**) and cardiac computed tomography (**C**), a 3-dimensional model (**D**, **E**, and **F**) was printed to plan the intervention. ASO indicates Amplatzer septal occluder; IVC, inferior vena cava; LA, left atrium; LUPV, left upper pulmonary vein; MV, mitral valve; RA, right atrium; and RV, right ventricle.

left atrial wall across the unroofed CS and deployed under TEE and fluoroscopic guidance with adequate closure of the defect. However, the device destabilized and prolapsed into the right atrium upon push-pull testing and was hence removed. A 30 mm Amplatzer septal occluder device was then deployed across the unroofed CS as previously simulated on our 3D model. TEE demonstrated minimal device interaction with the posterior mitral valve leaflet and unchanged mild mitral regurgitation. Push-pull technique confirmed stability of the device, and it was hence released from the delivery cable. TEE after release confirmed appropriate positioning of

Table. Hemodynamics and Oxygen Saturations at Baseline and After Deployment of 30 mm ASO Device at the Unroofed Coronary Sinus

	Baseline		After deployment of 30 mm ASO device	
Site/measurement	Pressure, mm Hg	O_2 saturation (%)	Pressure, mm Hg	O_2 saturation (%)
Inferior vena cava	12 (mean)	80%	11 (mean)	72%
Superior vena cava	12 (mean)	78%	11 (mean)	77%
Right atrium	12 (mean)	83%	10 (mean)	82%
Right ventricle	48/15		40/12	
Pulmonary artery	45/24 (mean 32)	86%	38/20 (mean 27)	81%
Left upper pulmonary vein	15 (mean)	93%	14 (mean)	95%
Descending aorta	117/69 (mean 87)	93%	117/66 (mean 84)	93%
Pulmonary blood flow, L/min	14.5		10.3	
Systemic blood flow, L/min	5.8		7.6	
Shunt fraction (Qp/Qs)	2.5:1		1.4:1	
Pulmonary vascular resistance, Woods Units	1.2		1.3	
Systemic vascular resistance, Woods Units	10.9		10.4	

ASO indicates Amplatzer septal occluder.

the device (Figure 2, Movie I in the Data Supplement) and no obstruction to pulmonary venous return, mitral, or CS ostium flow after the procedure (Movie II in the Data Supplement). Hemodynamic catheterization performed immediately after device deployment showed a marked improvement in the shunt fraction to 1.4:1, reduction in mean pulmonary artery pressure from 32 to 27 mm Hg, mild reduction in right-sided filling pressures and no increase in pulmonary vein pressure (Table). The patient was discharged home the following day on aspirin and clopidogrel. At 1-month follow-up, he was feeling well and reported significant improvement of his dyspnea.

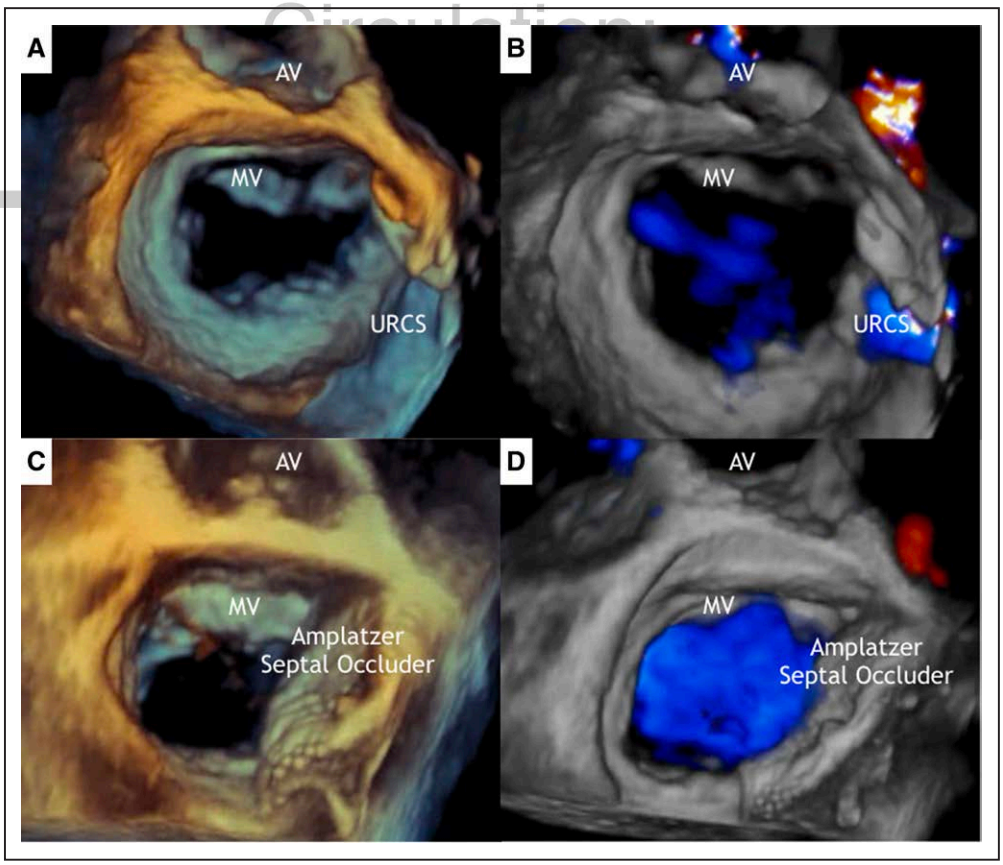

Figure 2. Three-dimensional (3D) transesophageal echocardiography images of the left atrium and mitral valve in early diastole. Images shown with and without color Doppler preintervention (**A** and **B**) and postintervention (**C** and **D**). AV indicates aortic valve; MV, mitral valve; and URCS, unroofed coronary sinus.

Unroofed CS defects are an exceedingly rare cause of pulmonary arterial hypertension. They account for <1% of atrial septal defects and have a strong association with a persistent left superior vena cava.[1] Surgical closure is the mainstay of treatment for unroofed CS defects and has been reported to have 3% in-hospital mortality in a single-center series of 159 patients.[1] However, in patients with a strong preference to avoid open surgery and with suitable anatomy, percutaneous closure has been shown to be safe and feasible.[2]

Our case demonstrates the advantages of using 3D printing, an emerging advanced imaging modality, to plan complex structural heart interventions. First, given the large size of the defect, the model enabled us to correctly identify the device sizes that would achieve adequate closure; this was verified by balloon sizing in the catheterization laboratory. Second, the model enabled us to size the device optimally to avoid mitral valve impingement, which is an important consideration given that the significantly larger left atrial disk of the device is positioned in the inferoposterior aspect of the left atrium, within close proximity of the posterior mitral valve leaflet. Third, in the case of a completely unroofed CS, this was especially useful in assessing the feasibility of alternate closure methods, such as a covered stent across the CS, which has been previously described.[3] Our patient uniquely lacked a persistent left superior vena cava draining into the left atrial aspect of the CS. As a result, the left atrial segment of the CS was smaller and tapered compared with the distal right atrial segment, which we noted on our 3D model. Therefore, we concluded that a covered stent would be difficult to size and appose and that a septal occluder device had a higher likelihood of success.

In this report, we highlight the importance of multimodality imaging and 3D printing for structural interventional planning. Important information such as device selection, sizing, and interaction with the posterior mitral valve leaflet can be obtained using 3D printing, setting the interventional team up for success and minimizing potential complications.

ARTICLE INFORMATION

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Disclosures

None.

Supplemental Materials

Suplemental Movies I–II

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